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CASE REPORTS THE DIAGNOSIS OF PONCET'S DISEASE

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SUMMARY

Two cases of polyarthritis in young people with extra-articular tuberculosis are described. The clinical features were consistent with the concept of Poncet's disease. After synovial biopsy one patient proved to have tuberculous peripheral arthritis. This observation raised doubts about the diagnosis in the other patient. Multiple joints may be simultaneously infected with *Mycobacterium tuberculosis* and Poncet's disease, if it exists, should not be entertained in the absence of synovial biopsy.

KEY WORDS: Polyarthritis, Tuberculosis, Synovial biopsy.

THERE is persistent scepticism about the existence of Poncet's disease, a sterile polyarthritis associated with non-articular tuberculosis [1]. There is even doubt about whether Poncet intended such an interpretation of his observations. His principal hypothesis implicated tuberculous infection (TB) in the aetiology of RA [2, 3]. We describe two patients with arthritis and TB who illustrate the uncertainties of Poncet's disease.

CASE REPORTS

Case 1

A 15-yr-old girl presented at our hospital with a 4-month history of neck swelling, joint pain and fever. She was otherwise well. Examination revealed bilateral cervical lymph node enlargement especially on the right (Fig. 1). The nodes were discrete and were not tender. There were no other abnormal findings apart from tender and warm swelling of the left wrist and fingers (Fig. 2), right elbow and ankle and the left knee. There was no demonstrable knee joint effusion. Investigations were Hb 11.1 g/dl, WBC $6.0 \times 10^9/l$, neutrophils 79%, ESR 95 mm/h, RF and anti-DNA negative. *Brucella* antibodies were negative and X-rays of involved joints were normal. A biopsy of one cervical lymph node confirmed the clinical suspicion of TB lymphadenitis by revealing typical granulomata. Culture did not yield mycobacteria. A rheumatologist suggested that the arthritis was due to Poncet's disease. A synovial biopsy was not performed and quadruple anti-tuberculous treatment was implemented. Within 4 months the lymph node enlargement and synovitis had resolved. Six months after commencing treatment she was well.

Case 2

An 18-yr-old male student was admitted to the hospital with an 8-month history of fever, neck, sternal and peripheral joint pain affecting the right shoulder, left wrist and ankle. His presentation coincided with that of case 1. There was no antecedent medical history. He was afebrile and there was pronounced restriction of head rotation. Right shoulder movement was limited by pain and the left wrist and ankle

were warm, swollen and tender. There was no neurological deficit. Investigations revealed: Hb 12.1 g/dl, WBC $7.0 \times 10^9/l$, neutrophils 70%, ESR 95 mm/h. *Brucella* antibodies were negative. X-rays of involved peripheral joints were normal but views of the spine demonstrated destruction of C3, 5, 7, D1 and L5 vertebrae with little loss of intervertebral disc spaces (Fig. 3). Computerized tomography was used to obtain a needle biopsy of the involved lumbar vertebra. This yielded tissue containing granulomata typical of TB infection. A diagnosis of TB osteomyelitis was made and quadruple anti-tuberculous treatment commenced. A rheumatologist suggested that the peripheral arthritis was due to Poncet's disease. An open synovial biopsy of the left wrist was obtained 5 days after treatment was started when the synovitis appeared to be receding. This revealed multiple caseating granulomata. The patient remained in hospital for several weeks, during which time the arthritis resolved and his neck pain improved sufficiently for him to be discharged wearing a collar.

DISCUSSION

Since 1974, there have been at least 11 English language reports of Poncet's disease describing a total of 19 cases [3–13]. Seven of these have derived from western countries where TB is now uncommon. In Pakistan, the infection is endemic but as elsewhere, skeletal and joint involvement is the least common manifestation [14]. A recent survey spanning 10 yr described 52 cases of peripheral TB arthritis [15]. None was ascribed to Poncet's disease even though 10% had at least two joints involved simultaneously. The report confirmed that young patients are especially susceptible to articular disease and that oligoarthritis with involvement of the upper limbs is more common than is supposed. The notion that TB arthritis is exclusively monarticular is a misconception. Emphasis of this characteristic has been deployed to support the diagnosis of Poncet's disease in polyarticular presentations [5]. In two such instances, an affected joint subsequently proved to be a focus of infection [3, 7].

Critical examination of most reports of Poncet's disease reveals anomalies that raise doubts about the attribution of arthritis. The acceptance of polyarthra-

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FIG. 1.—Case 1 showing right sided cervical lymph node enlargement.

lgia without objective evidence of synovitis has been an act of faith by some authors [2, 9]. Of the 19 cases described, synovial biopsy was undertaken in only four [3, 7, 8, 13]. All failed to reveal evidence of TB but biopsy of one joint was followed by subsequent isolation of mycobacteria [3] indicating that even biopsy may be inconclusive of TB arthritis. In our case 2, the clinical diagnosis of Poncet's disease was revised after open biopsy revealed the true nature of the arthritis. The patient also had infection of several vertebral sites, further evidence of the disseminating proclivity of the organism. It is of interest that two previous reports of Poncet's disease also had spinal osteomyelitis [8, 13]. These, like both our patients were young. Of all the quoted cases, only three were aged more than 40 yr

[11, 12]. It is noteworthy that it is the younger age groups which appear more likely to develop TB infective arthritis [15].

Our Case 1 resembled examples of Poncet's disease described by some others in as much as she had objective involvement of more than three joints [8, 13]. The tender swelling of the fingers was probably secondary to wrist synovitis. She did not undergo synovial biopsy in deference to her age and the rheumatologist's diagnosis. However, Case 2 manifestly confirmed that multiple joints may be infected simultaneously by *Mycobacterium tuberculosis* and the experience made the diagnosis of Poncet's disease in Case 1 less secure. The arthritis of both patients responded to anti-TB treatment and it is therefore likely that the arthritis was



FIG. 2.—Swollen left wrist and fingers in Case 1.



FIG. 3.—Cervical spine X-ray of Case 2 revealing tubercular osteomyelitis at C3 and C5 vertebrae.

not coincidental to tuberculosis. This is the usual therapeutic outcome described for Poncet's disease which unlike reactive arthritis has been associated only with continuing active infection at extra-articular sites.

The possibility of Poncet's disease representing an immune response to mycobacterial antigens has, despite the apparent rarity of the disorder, prompted speculation about the relationship between arthritis, 65 kD heat shock protein, T cell responsiveness and immunogenetic susceptibility [16, 17]. Although such speculation is of interest it would be advisable if future descriptions of Poncet's disease offered for publication were scrutinized with exceptional care. No case should be accepted as further proof of such an entity without evidence of rigorous exclusion of TB joint infection

including synovial biopsy. The apparent paucity of the condition in areas of the world where TB is common and recognition that the organism may occasionally infect several joints makes us sceptical about the concept of Poncet's disease as a polyarticular immune response to *Mycobacterium tuberculosis*.

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